

Benign Splenic Epithelial Cyst Accompanied by Elevated Ca 19-9 Level:

A Case Report

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Abstract

We report the case of a 30-year-old woman with a benign epidermoid splenic cyst and a high CA 19-9 serum level (268 U/mL). The patient underwent resection of the cyst and splenectomy. After removal of the cyst, the serum CA 19-9 level decreased to a normal level within 6 weeks. True non-parasitic splenic cysts are rare. Approximately 30 cases of benign true splenic cysts with a high CA 19-9 serum level have been published.

Key Words: Splenic epidermoid cyst, CA 19-9.

Introduction

CYSTIC CHANGES OF THE SPLEEN are very rare. Based on the presence or absence of cellular lining of the cystic wall, splenic cysts are classified as primary (true) or secondary (pseudo)cysts (1, 2). Primary splenic cysts other than those of hydatid disease, which is endemic in Turkey, are also very uncommon. So far, elevated carbohydrate antigen 19-9 (CA 19-9) serum levels accompanying splenic cysts have been reported in approximately 30 cases, of which most were of epithelial origin (3–20).

In this paper we report an additional patient with cystic lesion of the spleen in the left upper quadrant, accompanied by increased serum CA 19-9 level.

Case

A 30-year-old female patient with left upper quadrant and left shoulder pain, and mass in the left hypochondrium, was admitted to our hospital.

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The patient's medical history was unremarkable, and she denied any trauma.

On physical examination, a palpable mass was found in the left upper quadrant. Laboratory data were normal except for high CA 19-9 serum level (268 U/mL; normal: 0–27 U/mL). The level of other tumor markers, such as CEA and CA 125, was also normal. Serologic tests were negative for hydatid disease. Abdominal ultrasound and computed tomography revealed a splenic cyst 10 cm in diameter; the cyst was apparently in direct contact with the stomach, spleen, and pancreas. No other intra-abdominal abnormality was found (Fig. 1).

Exploratory laparotomy confirmed a centrally located splenic cyst 10 cm in diameter. No other pathology was detected. Splenectomy was per-

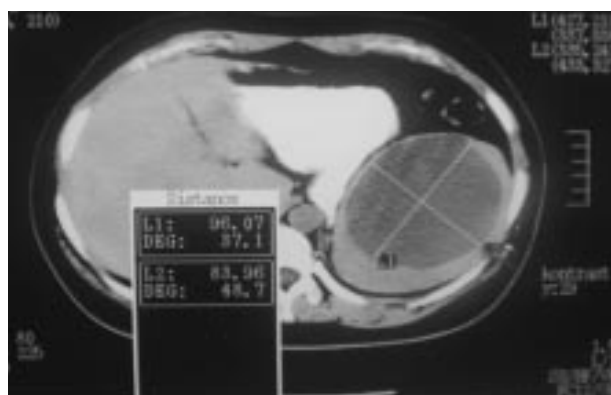


Fig. 1. CT scan showing a large and centrally located splenic cyst.

formed, since conservative treatment is not possible when the spleen is almost totally occupied by the cyst (Figs. 2A and B). The histopathological diagnosis was an epithelial splenic cyst (Fig. 3). The postoperative course was uneventful and the patient was discharged on the third postoperative day. CA 19-9 serum level returned to normal (8.9 U/mL) six weeks later. The patient was doing well, with normal CA19-9 levels, during a follow-up period of 30 months.

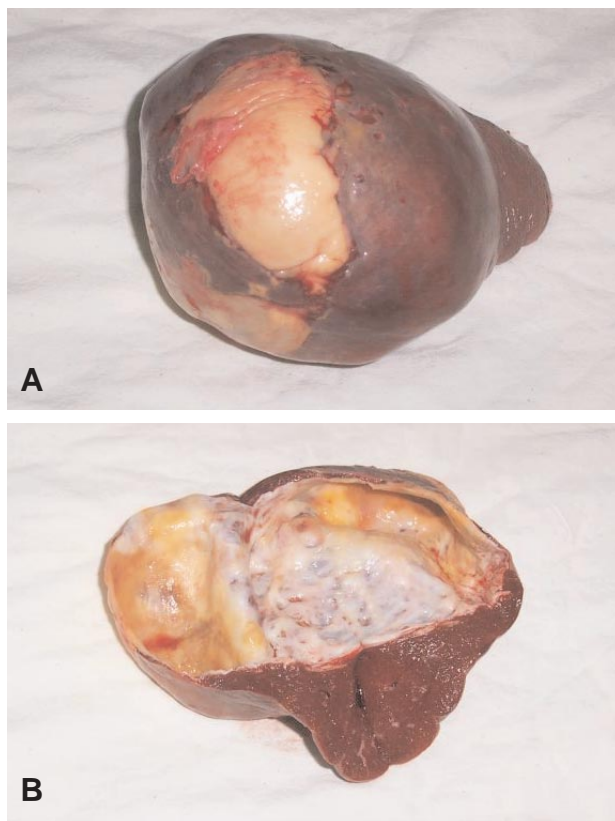


Fig. 2. (A) Macroscopic view of the splenic cyst. (B) Inside of the cyst.



Fig. 3. Hematoxylin-eosin stain of a section of the cyst wall showing epithelial cellular lining (H&E \times 40).

Discussion

Splenic cysts may be primary (true) or secondary (pseudocyst), depending on the presence or absence of an inner cellular lining (1, 2). Most true splenic cysts are epithelial in origin and have embryonic inclusion of epithelial cells from adjacent structures (6, 11, 21).

Although CA 19-9 is a reliable marker for pancreatic adenocarcinoma, both normal and tumoral human epithelial cells may produce it. High serum levels of CA 19-9 may accompany benign gastrointestinal diseases such as liver cirrhosis, acute cholangitis, obstructive jaundice and benign pancreatic conditions (6, 22, 23).

The first case of a splenic cyst with elevated serum CA 19-9 was reported by Terada et al. (20) in 1994. Since then, elevated CA 19-9 serum levels have been reported in approximately 30 cases of splenic cysts, most of them of epithelial origin (4–20).

Splenic epithelial cysts occur predominantly in children and young women (13). Small cysts are usually asymptomatic. The initial symptoms and signs referable to large cysts may include vague abdominal pain and a palpable mass in the left upper quadrant with or without symptoms due to compression of adjacent organs (10, 24).

Surgery is the only treatment for a splenic cyst with increased CA 19-9 level. In addition to conventional splenectomy, laparoscopic splenectomy and excision of the cyst and conserving spleen have been reported as alternative methods (13, 15).

Conclusion

Complete removal of the cyst by open or laparoscopic surgery is mandatory to ensure entire extirpation of cancer antigen-producing epithelium (6). Epithelial lining of true splenic cyst may be demonstrated by immunohistochemical analysis to produce this cancer antigen. Following removal of the cyst, serum levels of tumor markers should normalize (6, 20).

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